Cutaneous Leishmaniasis: Report of Two Atypical Cases

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Abstract: Multiple and unusual clinical features of cutaneous leishmaniasis (CL) had been reported as hyperkeratotic, psoriasiform, eczematoid, zosteriform pattern, warty lesions, erysipeloid, and acneiform lesions. The clinical features of CL primarily depend on the host's cell-mediated response and the species of *Leishmania* being involved. In this paper we reported two cases of atypical CL from Iran, Shohada-e-Tajrish and Loghman-e-Hakim hospitals presented with infiltrative erythematous lesions covering the perioral region and the lower limb, respectively. The diagnosis of cutaneous leishmaniasis was confirmed with skin smears and biopsies.

Keywords: New world leishmaniasis, Old world leishmaniasis, Cutaneous leishmaniasis, leishmaniasis, Atypical cutaneous leishmaniasis.

INTRODUCTION

Cutaneous leishmaniasis (CL) is a parasitic infection encountered in daily dermatologic practice [1]. Iran has high rate of this widespread tropical infection [2]. Based on the geographic region where the infection is acquired, it is divided into two subsets: old world and new world. The causative organisms, vectors, reservoirs, clinical manifestations, and prognosis are different in these groups. Usual causes of old world CL are due to *L. major* or *L. tropica* [3]. The main reservoirs of this disease are infected patients and dogs [1]. The two forms usually begin as a small papule at the inoculation site; which gradually enlarges over several weeks into a nodule and then becomes ulcerated or verrucous. Exposed sites are most commonly involved. The majority of these lesions will resolve spontaneously with scar formation [3]. Atypical clinical presentations have been reported which include hyperkeratotic, psoriasiform, eczematoid, zosteriform pattern, massive auricular enlargement, diffuse cutaneous leishmaniasis with lymphoma like lesions, warty lesions, erysipeloid, and acneiform lesions [4-8]. Erysipeloid form is an uncommon manifestation of CL. This atypical clinical feature has been reported from Iran, Turkey, Pakistan and Tunisia [5, 9-11].

CASE PRESENTATION

The first case was a 25-year-old man presented with a crusted, infiltrative and erythematous plaque on his perioral region. His history revealed that the lesion started from an asymptomatic and ulcerated papule on the upper labial region, then slowly enlarged and involved his chin and perioral region (Figure **1**). He had no significant past medical history except for traveling to an endemic area of CL in Iran. No lymphadenopathy, mucous membrane involvement, or systemic abnormalities were detected. Routine laboratory results were normal. A skin smear taken from the lesion was positive for CL with many amastigotes in the intracellular and extracullar area of the macrophages(Figure **2**).

Figure 1: (a), (b) A crusted and erythematous plaque on the chin and perioral region.

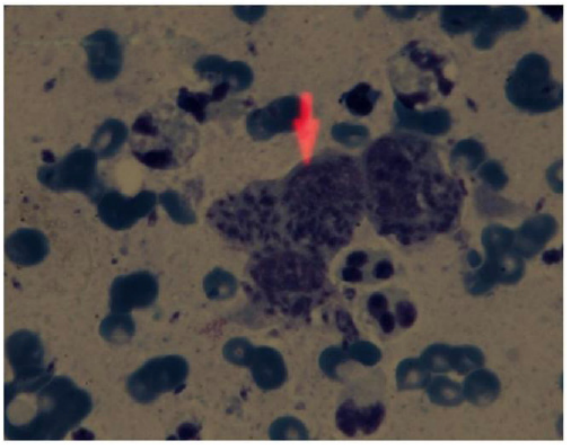

Figure 2: The skin smear shows many amastigotes in the intracellular and extracellular area of the macrophages (Geimsa stain, × 100).

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Figure 3: (a) Multiple erythematous indurated papules and plaques on the anterior aspect of the left leg and these lesions spread to his left toes. **(b)** Also an ulcer with erythematous margin existed on the third toe of the left foot.

The second case was a 40-year-old man presented with multiple erythematous indurated papules and plaques on his left shin, ankle and foot and ulcerative lesions on the posterior and anterior aspects of his left shin. He had traveled to an endemic area of CL in Iran. Also an ulcer with erythematous margin was observed on the third toe of the left foot (Figure **3**). Physical examination of the mucous membranes and other organs was normal. The skin smear was positive for *leishmania* and the histopathologic examination of skin biopsy was positive for CL (Figure **4**).

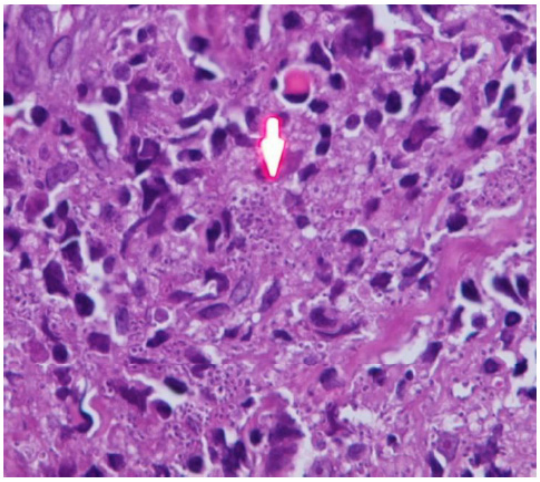

Figure 4: Histopathologic examination of the skin biopsy shows numerous amastigotes in macrophages (H & E, × 40).

Both patients were treated intramuscularly with 10 mg/kg/day systemic meglumine antimoniate. After 20 days of systemic meglumine antimoniate therapy, marked improvement of lesions were observed. Two weeks after treatment, only scars were observed in the place of the ulcerated lesions (Figure **5**).

Figure 5: case 1: after treatment with systemic meglumine antimoniate.

DISCUSSION

CL is one of the most important zoonotic diseases in Iran, and it is endemic problem in South and Central part of Iran. *L.tropica* and *L.major* cause this disease in Iran [2, 5]. It usually begins as a small papule which may gradually enlarge over several weeks into a nodule and become ulcerated [3]. This disease shows various symptoms depending on host's cell- mediated immune system. The reasons for atypical presentation such as erysipeloid form are unclear, although alteration in host's immune response, involvement of specific subtypes of parasite, changes in hormones and skin barrier can be considered as important factors [12].

Erysipeloid type of CL was first reported from Iran in 1994 on the face of predominantly female patients [5]. In 1998, an erysipeloid case presented as an erythematous, indurated plaque over the right side of

the upper lip and adjacent cheek was reported [9]. In 1999, Salmanpour *et al* reported five Iranian patients, predominantly females (50-70 years of age) presented with infiltrative erythematous lesions on the center of the face [12]. In 2004, Karincaoglu described an erysipeloid case of CL in a Turkish woman, presented with butterfly-shaped infiltrated erythematous plaque on the face, [10] and Ceyhan and colleagues reported a similar case that presented with ulceration on glabella and infiltrative erythematous lesions covering the center of the face. The diagnosis can be confirmed by the presence of amastigotes in dermal macrophages of skin biopsy, but Giemsa staining is used to detect leishmania in smear [1].

The differential diagnosis includes fungal infections, eczema, leprosy, atypical mycobacterial infections, sarcoidosis, lupus vulgaris, basal and squamous cell carcinomas, tuberculosis, infected insect bites and cutaneous metastasis of internal malignancies [13, 14].

Treatment depends on the region of the world in which the infection was acquired, the species of *Leishmania*, the site(s) and severity of the infection, and host factors such as immune status and age. Old world cutaneous leishmaniasis is often self-limiting and without treatment, typically resolves within 2-4 months (*L. major*) or 6-15 months *(L. tropica)*. Pentavalent antimonials as sodium stibogluconate and meglumine antimoniate are standard treatment for cutaneous and mucocutaneous leishmaniasis. Cryotherapy, heat therapy (to 40-42°C for several hours each day), paromomycin (topically BID × 10-20 days), miltefosine (2.5 mg/kg/day PO (max150 mg) × 28 days), itraconazole (7 mg/kg/day PO × 3 weeks), pentamidine (2-3 mg/kg IV or IM daily or every other day × 4-7 doses), amphotricine B (preferred treatment for visceral type), alluporinol (10-20 mg/kg for 6 to 8 weeks), ketoconazole (600 mg/day or 10 mg/kg/day PO × 4 weeks) are additional therapies used to treat CL. Plastic repair may be used for severe scarring [3, 15].

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